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Article in *Surgical Neurology International* · December 2020

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Case Report

Painful torticollis due to tubercular atlantoaxial rotatory fixation: A case report

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Received : 20 September 2020

Accepted : 25 November 2020

Published : 16 December 2020

DOI

10.25259/SNI_663_2020

Quick Response Code:



ABSTRACT

Background: Tubercular atlantoaxial, rotary dislocation warranting fixation (AARF) is an extremely rare event.

Case Description: AARF was suspected in a 23-year-old female with painful torticollis. When diagnostic studies documented unilateral destruction of the left lateral mass of the atlas, she underwent removal of the lateral mass, reduction of the deformity, and C1-C2 fusion/reconstruction utilizing an iliac bone graft. Laboratory tests and the pathologic surveys were all consistent with the diagnosis of underlying tuberculosis.

Conclusion: We present a case of tubercular atlantoaxial, rotary dislocation (AARF) in a patient who warranted C1-C2 decompression, reduction, and fusion.

Keywords: Atlantoaxial dislocation, Atlantoaxial rotatory fixation, Atlas, Craniovertebral junction, Tuberculosis, Upper cervical

INTRODUCTION

Tuberculosis (TB) involving the atlantoaxial complex is rare, accounting for approximately 0.1% of all spinal tubercular infections.^[1,3,5,11,14] Here, we present a young woman with painful torticollis attributed to tubercular atlantoaxial rotary dislocation (AARF).

CASE REPORT

A 23-year-old female presented with a 4-week history of severe neck pain (VAS:10) and torticollis with a classic "Cock Robin" deformity. Laboratory studies demonstrated an increased erythrocyte sedimentation rate (ESR) of 42. The lateral cervical plain radiographs and computed tomography (CT) axial, 2D, and 3D reconstructed images showed C1-C2 AARF with destruction of the left lateral mass of the atlas [Figures 1-4]. The integrity of the vertebral artery at C1-C2 was also critically confirmed on the preoperative CT angiogram (CTA) [Figure 5].

Surgery

As cervical traction failed to reduce the deformity, surgical intervention was warranted. Surgery required; the initial insertion of bilateral C2 pedicle screws, isolation of the V3

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segment of the vertebral artery, removal of the destroyed left C1 lateral mass in a piecemeal fashion, and fusion (e.g., utilizing a tricortical iliac bone graft secured with left C1 laminar hook-C2 pedicle screw, and an additional right C1 lateral mass-C2 pedicle screw rod construct) [Figure 6]. Postoperatively, the patient's torticollis and intractable pain resolved. X-rays taken 1 week, and 3 months after surgery demonstrated adequate alignment of the instrumentation/construct [Figure 7]. Three years later, the patient is asymptomatic [Figure 8].



Figure 1: Lateral cervical radiograph showing C1 foramen transversarium indicating possible rotation of atlas (white arrow).

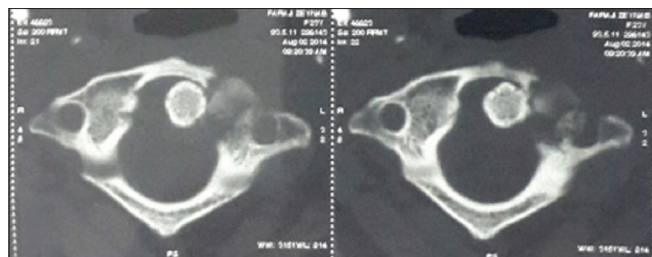


Figure 2: Axial computed tomography scan showing destruction of the left C1 lateral mass, note approximation of the odontoid to the left lateral mass.

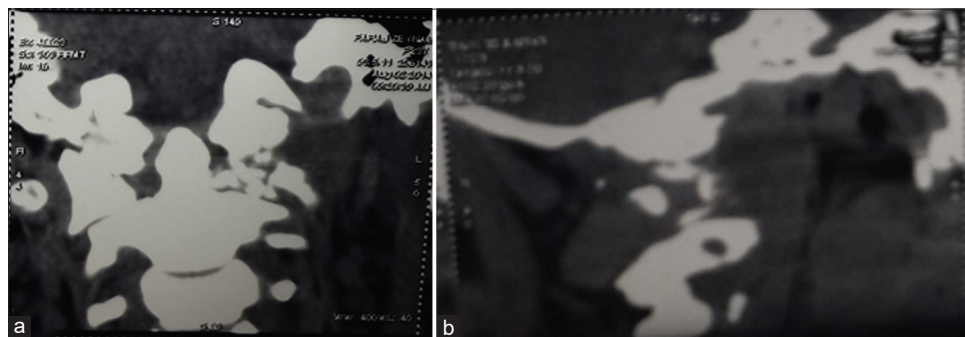


Figure 3: Reconstructed computed tomography scan (a) coronal, (b) sagittal showing destruction of the left lateral mass of atlas.

Bacteriology and pathology

The operative specimens demonstrated: a positive polymerase chain reaction (PCR) for TB, and the pathology was compatible with a granulomatous infection.

Tubercular treatment

Four-drug therapy was warranted for TB; isoniazid (5 mg/kg), rifampicin (10 mg/kg), ethambutol (15 mg/kg), and pyrazinamide (25 mg/kg). They were administered as a first-line of treatment for 4 months. This was followed by an additional 12 months of rifampicin and isoniazid.

DISCUSSION

Spinal TB, presenting as AARF with painful torticollis, is extremely rare.

Pathogenesis

Severe painful torticollis may be the only clinical indication that AARF is present. Patients may exhibit unilateral destruction of the lateral mass of the atlas with/without infiltration/disruption of the alar ligaments. The ESRs are typically increased, and the Mantoux test is typically positive.^[1-5,9,11,14] Further, acid-fast stains of pathological material, positive cultures, and PCR obtained through CT-guided biopsy may be additional valuable adjuncts to confirm the diagnosis of TB.^[1-5,9,11,14]

Imaging

AARF is the best documented utilizing axial reconstructed 2D and 3D CT images and CTA.^[12]

The following findings are classical for TB; an osteolytic, fragmented lesion involving the C1 lateral mass, deviation of the odontoid to the affected lateral mass side, and forward displacement of the contralateral lateral mass of atlas.^[4,6,7,10,13,15] Magnetic resonance imaging also may help to establish the diagnosis of tuberculous involvement of the C1 lateral mass,

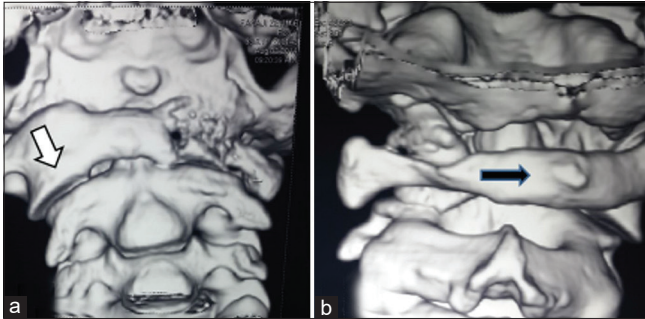


Figure 4: 3D computed tomography scan of the atlantoaxial complex (a) frontal view showing destruction of the left lateral mass and a part of anterior ring of atlas, note forward displacement of the right C1 lateral mass. (b) Occipital view demonstrates rotation of the posterior ring of atlas to the right.

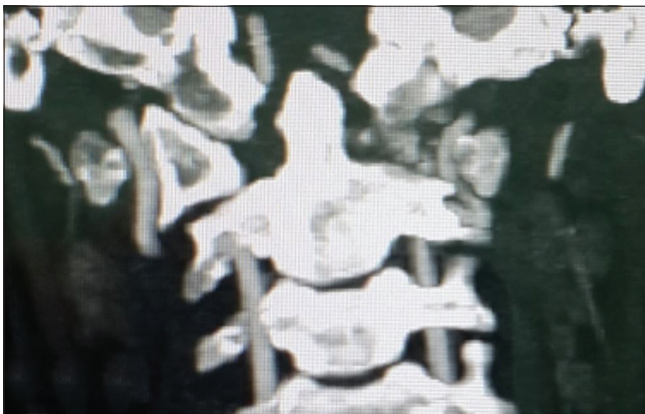


Figure 5: Reconstructed coronal computed tomography angiography shows integrity of the left V3 segment of the vertebral artery passing through the destroyed lateral mass.

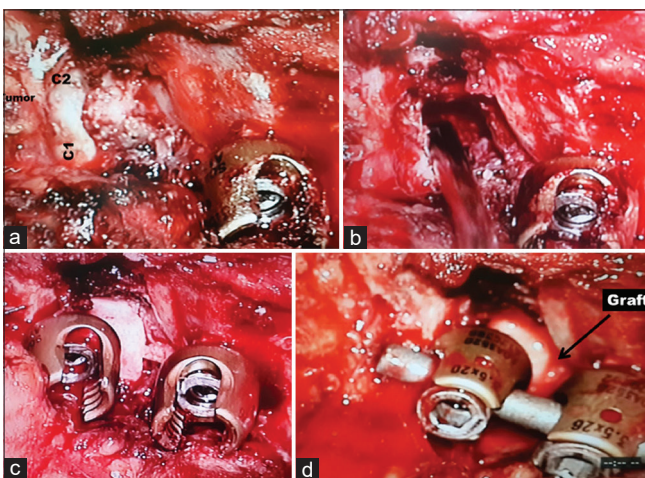


Figure 6: Intraoperative photograph, (a) the lesion of the left lateral mass of atlas. (b) The site after removal of the mass. (c) The lateral mass is reconstructed with tricortical iliac autogenous bone graft, note C2 pedicle and C1 hook. (d) After assembling the rod.



Figure 7: Postoperative X-ray. (a) A week after surgery showing the C2 pedicle-C1 lateral mass and hook construct used properly. (b) At 1-year follow-up.



Figure 8: Photograph of the patient, a year after surgery while holding up her cervical radiographs.

by demonstrating heterogeneous intensity on the T1, and hyperintensity on the T2-weighted and fat-suppressed images.

Differential diagnosis

The differential diagnoses for painful torticollis with unilateral involvement of the C1 lateral mass include TB tumors, rheumatoid arthritis, and other types of pyogenic spondylitis.^[4,6,7,8,10,13,15]

Management

In classic tubercular atlantoaxial dislocation, management strategies range from purely conservative treatment to radical operations.^[1,5,11,14] Surgical intervention for decompression, reduction/realignment, and instrumented fusion may also be warranted.

CONCLUSION

Here, we presented an extremely rare cause of painful torticollis due to tubercular AARF involving a unilateral C1 lateral mass requiring decompression, reduction, and fixation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Rahimizadeh A, Williamson W, Rahimizadeh S, Amirzadeh M. Painful torticollis due to tubercular atlantoaxial rotatory fixation: A case report. *Surg Neurol Int* 2020;11:440.